Carcinoma of the Thyroglossal Duct

Primary or Metastatic from the Thyroid?

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TWENTY-FOUR CASES of carcinoma of the thyroglossal duct have been reported.[8] In all but two cases[1,5] the lesions were papillary adenocarcinomas, and in only two was there demonstrable metastatic tumor in regional lymph nodes.[3,4] None of the patients died directly from recurrence or spread of the carcinoma. Surprisingly, only two[1] of the reported patients were treated with concomitant thyroidectomy.[7,8] In no previous case was an associated carcinoma of the thyroid gland reported.

In the present case, an occult papillary adenocarcinoma of the thyroid was presumed to be a separate primary neoplasm and, as such, aroused special interest and speculation regarding therapy.

Report of a Case

The patient, a 23-year-old woman, was admitted to hospital 20 February 1966 because a firm, submental mass that had been present for as long as the patient or her family could remember had doubled in size—to 6 cm—within the preceding year. There was no history of inflammation.

On physical examination the mass could not be moved laterally but it moved up and down with swallowing. The surface was irregular, with one firm nodule 1 cm in diameter. The thyroid gland was small, symmetrical and smooth, without nodules. No other masses were palpable within the neck.

I^{131} uptake was 53 per cent in 24 hours, and an I^{131} scan showed uniform activity within the thyroid gland. There was no uptake outside the thyroid gland or over the submental mass.

At operation the mass was exposed with a long, transverse, elliptical incision and was found to be intimately fixed to the surrounding thyrohyoid, sternohyoid, digastric and mylohoid muscles. Without biopsy of a specimen the mass was excised in toto, including the muscles to which it was fixed and a 1-inch segment of the central portion of the hyoid bone. The pathologist* reported a frozen section showed mixed papillary and follicular carcinoma of the thyroid, infiltrating skeletal muscle. However, there also was a benign epithelial element, interpreted as showing origin within a thyroglossal duct cyst. (See Figures 1, 2 and 3).

An occult primary tumor within the thyroid gland was considered an outside possibility, inasmuch as no nodularity of the thyroid was palpable and the I^{131} uptake was uniform. However, it was felt desirable to ablate the thyroid totally, so that any recurrence might be identified by I^{131} uptake and then destroyed with I^{131} therapy. Total thyroidectomy was undertaken ten days after the initial procedure. No nodularity was palpable when the gland was exposed, and the only lymph nodes encountered in the sugical procedure were negative for tumor. However, on subsequent step cuts of the totally embedded thyroid gland, the pathologist† reported a 4 mm primary mixed follicular and papillary carcinoma (Figure 4).

Postoperatively, the small remnants of functioning thyroid tissue were ablated with I^{131} and thereafter the patient was given 0.26 gm of thyroid extract daily. It is planned that periodic scanning with I^{131} for any functioning thyroid tissue will be

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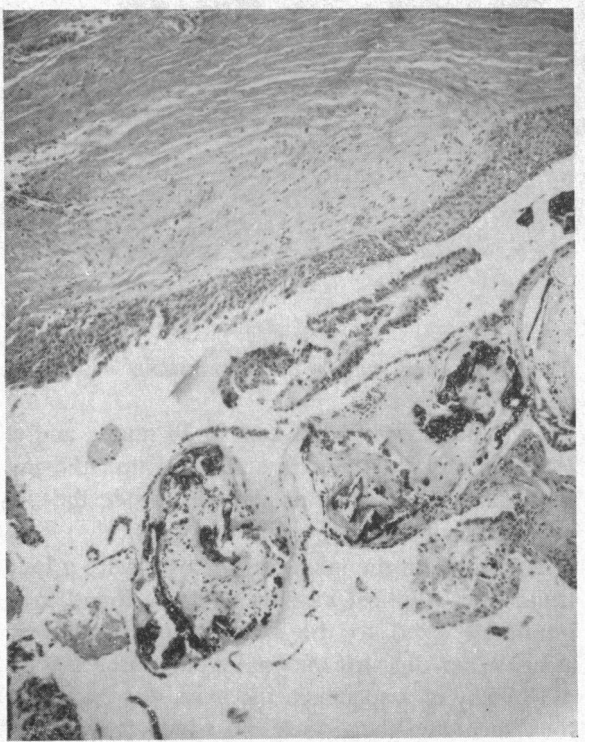

Figure 1.—Thyroglossal duct cyst wall and surrounding hyalinized papillary carcinoma (× 100).

carried out and any functioning thyroid again ablated with I^{131} (to date, two years after operation, there is no evidence of recurrence).

Discussion

Any rapid increase in size or a very large, nodular, fixed, submental mass may give rise to suspicion of carcinoma within a thyroglossal duct cyst. Failure of the mass to show any function with I^{131} scanning does not rule out the possibility of thyroid carcinoma within the thyroglossal duct cyst. If fixation or invasion of surrounding musculature is noted at operation, it would seem advisable to resect the lesion en bloc. However, most cases reported in the literature appeared to be "typical" thyroglossal duct cysts, and the correct diagnosis was made only after microscopic examination of the surgical specimen.

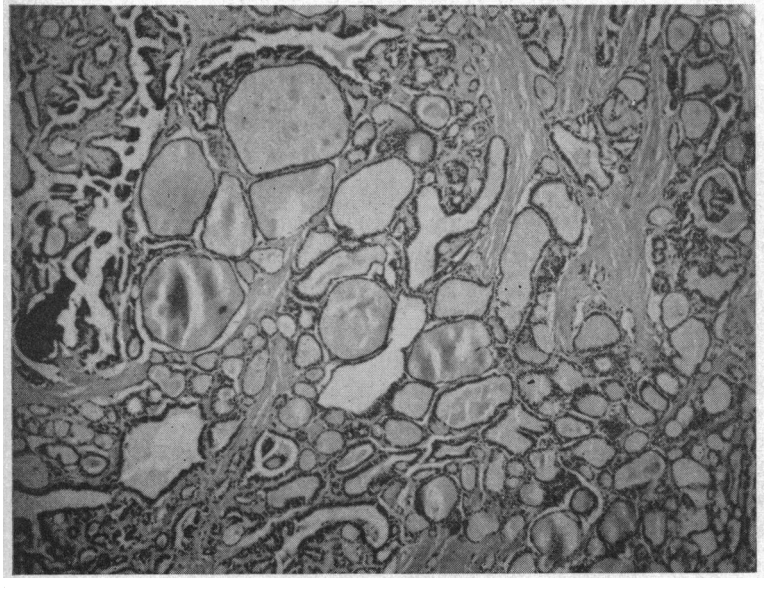

Figure 2.—Papillary and follicular thyroid carcinoma in thyroglossal duct remnant (× 100).

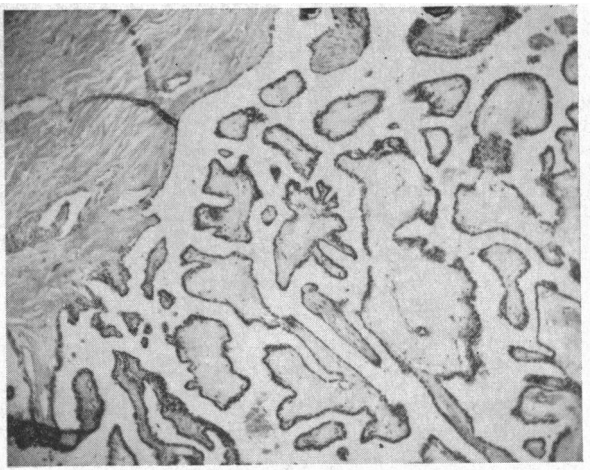

Figure 3.—Papillary carcinoma in thyroglossal duct remnant (× 100).

Although in the present case the interpretation was that there were two primary lesions, it is pertinent to note that Crile[2] said that the thyroglossal duct tract may be a route of spread of carcinoma of the thyroid. Nuttal[6] eloquently stated the possibility of confusing *cystic* metastasis from an occult

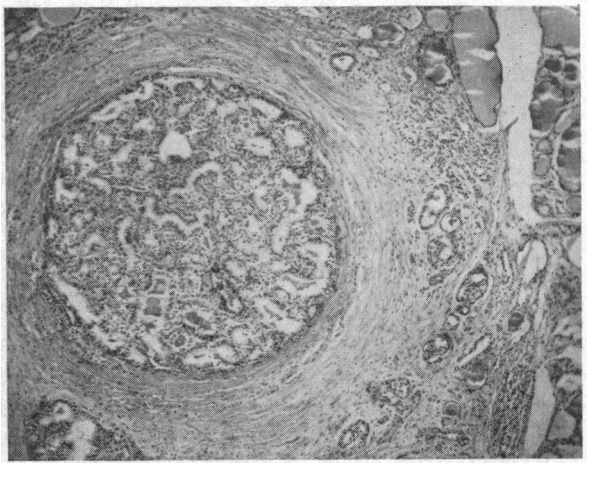

Figure 4.—Small primary papillary carcinoma in thyroid gland (× 100).

primary carcinoma of the thyroid gland with carcinoma originating within a thyroglossal duct cyst. Woolner and coworkers[9] reported upon two cases of occult papillary carcinoma of the thyroid in which a previous diagnosis of "carcinoma in thyroglossal duct cysts" had been made erroneously. On microscopic examination these cystic metastatic lesions, however, typically have a thin wall, with or without lymphoid tissue, which may be mistaken as evidence of thyroglossal duct cyst.

Some doubt may always remain whether the submental mass in the present case was a metastatic tumor. However, the history of the patient's having had a submental mass "all her life" and the microscopic evidence of a benign epithelial component are taken as strongly indicative that the submental carcinoma originated within a thyroglossal duct. One can speculate that whatever factor (perhaps excess thyroid stimulating hormone) stimulated carcinogenesis within the ectopic thyroid tissue may also have engendered the carcinoma within the thyroid gland itself.

Perhaps consideration should be given to total thyroidectomy in patients found to have thyroid carcinoma in a thyroglossal duct cyst, not only to rule out an occult primary lesion but also to make follow-up I^{131} scanning and ablation of recurrences possible. As the present case illustrates, an occult primary lesion may escape detection even on palpation of the thyroid gland at operation and gross examination by a pathologist. Thyroidectomy then would remain, in those circumstances, the only means by which an occult primary lesion could be demonstrated. Finally, suppressive doses of desiccated thyroid seem indicated.

Summary

The twenty-fifth reported case of carcinoma occurring within a thyroglossal duct cyst is reported. The presenting submental mass was somewhat atypical in that it was large—6 cm—and fixed because of invasion of surrounding musculature. This invasion made en bloc resection of these muscles necessary. A separate occult primary lesion was found within the thyroid gland. Total thyroidectomy seems advisable in the treatment of thyroid carcinoma occurring within a thyroglossal duct cyst, since the procedure would make subsequent I^{131} uptake studies, scanning and ablation of metastatic lesions possible.

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Prinzmetal's Variant of Angina Pectoris with Only Slight Coronary Atherosclerosis

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SINCE THE original report[3] of the syndrome which is now known as Prinzmetal's variant of angina pectoris, several observers[1,2,4] have published accounts of this interesting constellation of findings. Pathologic examinations in such cases have revealed decided arteriosclerotic narrowing of a large coronary artery.[3] In the case here reported, Prinzmetal's variant was associated with only mild atherosclerosis of the coronary arteries.

Arrhythmia frequently occurs during the is-

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